

## DESTRUCTIVE ASPERGILLOSIS

**Surinder K. Singhal<sup>1</sup>, Arjun Dass<sup>2</sup>, G. B. Singh<sup>3</sup>, R. P. S. Punia<sup>4</sup>, Nitin M. Nagarkar<sup>5</sup>**

**ABSTRACT:** The incidence of mycotic infections of paranasal sinuses is on the rise. Aspergillosis is the commonest fungal infection involving the nose and paranasal sinuses. This disease has a varied presentation ranging from the allergic form to the more deadly invasive or destructive form. Destructive fungal disease is almost regarded as synonymous with mucormycosis. The destructive potential of aspergillosis has not been highlighted in the medical literature. We here report a case of aspergillosis, which caused massive destruction of maxilla in an apparently healthy patient.

**Key Words:** aspergillosis; forehead flap; mycotic infections.

Mycotic infections of the paranasal sinuses are on the rise globally. This is probably attributed to the rise in the incidence of AIDS and drug abuse. Of all the fungal infections, aspergillosis is the commonest fungal disease in human beings.<sup>[1]</sup> Destructive, i.e. invasive form of aspergillosis in an apparently healthy patient, is still rare. What makes the disease sinister is its indolent course and ability to cause death. A fatality rate of 16% has been reported in the medical literature<sup>[2]</sup>. The disease has a good prognosis provided it is diagnosed early and treated properly with surgery and antifungal therapy.

In this paper, we report a case of invasive aspergillosis with massive destruction along with the historical perspective of the disease.

### **CASE REPORT**

A 51-year-old male farmer by occupation presented to us in the out patient department with a large defect in the right cheek and palate for the past 1 year. Six years back he started having repeated attacks of postnasal discharge and headache along with nasal obstruction off and on for which he received medical treatment. Later on the patient had loosening of right upper molar tooth, which was extracted. Thereafter, he developed a discharging tooth socket. He was then referred to another hospital, where he underwent Caldwell-Luc's operation and was diagnosed to be suffering from aspergillosis. He was put on antifungal treatment in the form of Amphotericin B & Itraconazole, but details of the treatments were not available. One year back he developed a discharging

sinus near the medial canthus on the right side and adjoining dorsum of nose, which gradually enlarged over a period of time and lead to the cheek defect [Figure 1]. There was no history suggestive of any systemic illness, apart from history of asthma for the last 12 years for which he was taking inhalational steroids irregularly.

On local examination, a defect of 6 cm x 3 cm in maximum dimensions was seen involving right cheek. Margins of the cheek defect were having granulation tissue with areas of slough and debris. Medial wall of the maxillary antrum was also destroyed and nasal septum was visible through the defect. There was a defect in the right half of the palate measuring 3 cms x 2 cm. The margins of the defect were



Figure 1: Clinical photograph of the patient showing defect in cheek and side of the nose.

<sup>1</sup>Senior Lecturer, <sup>2</sup>Prof. & Head, <sup>3</sup>Senior Resident, <sup>4</sup>Senior Lecturer, <sup>5</sup>Reader, Departments of E.N.T. and <sup>4</sup>Pathology, Government Medical College Hospital, Chandigarh, India.



Figure 2: CT scans of the patient axial section (A), coronal section (B) showing destruction of maxilla and palate.

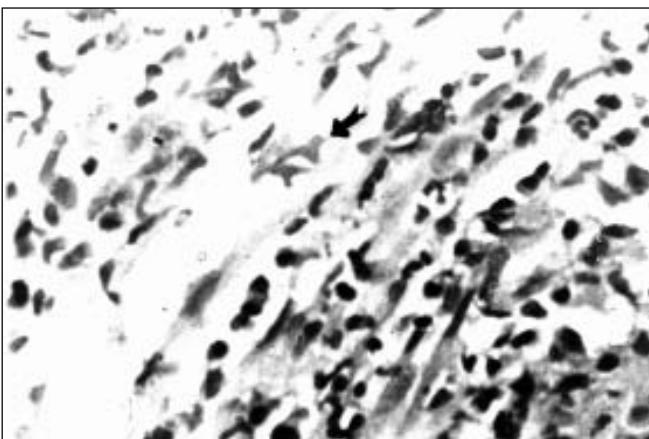


Figure 3: Photomicrograph showing septate hyphae of aspergillosis (arrow) H&E 200'.

healthy. The CT scan was done and showed destruction of maxillary sinus with loss of overlying skin and subcutaneous tissue with soft tissue involvement, palatal defect and lateral nasal wall destruction [Figure 2A and B]. Biopsy was taken from the granulation tissue at the margin of the defect in cheek, which revealed branching septate hyphae suggestive of aspergillosis [Figure 3] and was confirmed on Grocott's staining.

The patient was started on oral itraconazole 100 mg twice a day for a period of 3 months. Palatal prosthesis was given for the palatal defect. His hepato-renal functions were monitored. After 3 months of itraconazole, biopsy was taken again from the representative area and was found to be normal. The patient then underwent cheek reconstruction by midline forehead flap [Figure 3]. At follow up, 1 year after repair of cheek defect, the patient remains clinically free from the disease. [Figure 4]

## DISCUSSION

The Priest botanist Micheli first described aspergillosis in 1729.<sup>[3]</sup> Morrel Mackenzie in 1893, published the first report of aspergillosis of maxillary sinus.<sup>[4]</sup> Aspergillosis belongs to ascomycetes class and is a saprophyte. *Aspergillus flavus* is the species, which commonly affects paranasal sinuses in humans. Inhalation is the usual mode of infection but



Figure 4: Clinical photograph of the patient after reconstruction with midline forehead flap.

marijuana smoking and dental fillings are other modes of infections.<sup>[5,6]</sup> The disease is not contagious. The pathogenicity is attributed to its local invasiveness and types I and III hypersensitivity reaction.<sup>[1]</sup>

Clinically this disease presents as noninvasive, invasive, fulminant and allergic forms<sup>[8]</sup>. The first three forms are a continuous spectrum of the disease, with noninvasive at one end and invasive and fulminant at the other end. Noninvasive form usually presents in the form of refractory sinusitis whereas ocular and neurological complications are seen in invasive form. Aspergillosis has been associated with a fatality rate of 16%.<sup>[2]</sup> What causes the indolent form of the disease to become invasive and fulminant is not clear. Few predisposing factors have been proposed like longer duration of disease, improper treatment of noninvasive form, patient's immune status, local obstructive lesions of nose and paranasal sinus, site of disease especially involvement of sphenoid sinus and its close proximity to brain and allergy.

In an apparently healthy patient, local obstructive lesions of nose and paranasal sinuses are important factors. But this could not be verified retrospectively in our patient due to massive destruction. However, duration of the disease with its improper treatment might have played a significant role to transform the disease to invasive form.

All cases of suspicious aspergillosis should be subjected to radiology preferably CT scan. MRI is usually reserved for intracranial involvement. Even a plain X-ray can give useful contributory evidence of aspergillosis. A dense shadow akin metal has been described to be pathognomonic of fungal disease, especially in absence of foreign body. These dense shadows represent crystals of calcium phosphate and calcium sulfate, manganese, iron and magnesium.<sup>[9,10]</sup>

The definitive diagnosis, however, depends on tissue

examination showing septate hyphae with dichotomous branching and the characteristic aspergillosis fruiting head. Charcot laden crystals are seen in allergic type. Grocott's methenamine silver (GMS), Gridley and periodic acid Schiff are a few specific stains. The fungus can be cultured on Sabouraud's agar.

Each case has to be treated on its own merit, like in our case a cosmetic dimension was added for rehabilitation. In the coming millennium—with quality of life improving—more and more patients of invasive and destructive aspergillosis would certainly require cosmetic rehabilitation. However, it is best to classify patients into three groups<sup>[12]</sup> as has been described by Julian Rowe-Jones.<sup>[12]</sup> The first group comprising of noninvasive form includes allergic aspergillosis or aspergilloma. The second group comprises of semi-invasive aspergillosis, i.e. disease being locally destructive without tissue invasion. This group is best treated with surgical debridement and sinus ventilation followed and a course of itraconazole.<sup>[12]</sup> The third group comprises of the true fungal invasion either nonfulminant or fulminant form of disease and is treated with surgical debridement and sinus ventilation followed by a course of amphotericin B or flucytosine or a combination of both and thereafter a long-term course of itraconazole.<sup>[13]</sup>

Prognosis remains excellent in group A. Aggressive and proper management of patients in group A including diagnosis and treatment of local obstructive lesions of nose and paranasal sinuses would certainly decrease the mortality and morbidity rates by preventing the disease to transform to its invasive form.

## CONCLUSION

From this case report, we conclude that though it is a common impression that mucormycosis presents with destruction of tissues, aspergillosis can also present in destructive form. Destructive and invasive forms of aspergillosis can occur in an apparently healthy patient. Moreover, all cases of indolent and protracted aspergillosis have a potential to transform to

invasive from, thereby highlighting the importance of initial proper treatment of noninvasive form of the disease.

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## *Address for Correspondence*

Dr. Arjun Dass, DHM  
Prof. & Head,  
Department of ENT,  
Government Medical College Hospital,  
Chandigarh 160047,  
India, E-mail: arjundr@usa.net